A case report of Burning Mouth Syndrome treated with individualized Homeopathy

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ABSTRACT

Introduction: Burning Mouth Syndrome (BMS) is a rare disease of unknown origin. No efficient treatment is known, and integrative approaches are warranted. So far, individualized homeopathy (iHOM) has not been evaluated or reported in peer-reviewed journal as a treatment option.

Method: At a centre of complementary medicine at a University institute in Switzerland, a 38-year old patient with BMS and various comorbidities has been treated with (iHOM) between July 2014 and August 2018. The treatment is characterized by the prescription of individually selected homeopathic single remedies. During follow-up visits, outcome was assessed with two validated questionnaires concerning patient reported outcomes. In order to assess whether the documented changes are likely to be associated with the homeopathic intervention, an assessment using the modified Naranjo-criteria was performed.

Results: Over an observation period of four years, an increasingly beneficial result from iHOM was noted for oral dysesthesia and pains as well as for the concomitant symptoms.

Conclusion: Considering the multi-factorial aetiology of BMS, a therapeutic approach like iHOM which integrates the completeness of symptoms and complaints of a patient might be of value in cases where an association of psychological factors and the neuralgic complaints is likely.

Keywords: Burning Mouth Syndrome; Pain management; Integrative Medicine

INTRODUCTION

Patients with sore, burning, tingling or numb parts of the oral cavity are classified as having Burning Mouth Syndrome (BMS), when the oral examination is normal (1) and there are no secondary causes for the presenting symptoms (2). Primary BMS is a rare disease with an incidence ranging from 0.01-0.1% of the general population, with the highest incidence in postmenopausal women above 70 years of age (3). To date, origin and pathogenesis of BMS remain unknown. Mostly aetiology is referred to as multifactorial, though a neuropathic pathway involving the trigeminal nerve, chorda tympani and glossopharyngeal nerve is favoured (4-6). Psychological and personality factors may be linked to the occurrence of BMS as well (7). There are currently no universally accepted guidelines for managing BMS and in a recent literature review and meta-analysis, the results of various treatment options yielded a significant benefit over placebo concerning pain scores (8). Further, despite several therapeutic approaches it was found in a retrospective analysis of 48 women that the course of the disease varies from spontaneous complete remission (3%) to no improvement in close to 70% of the patients after five years (9). The authors conclude that new, safe and efficacious treatment options should be developed. As multiple factors are linked in the development and evolution of BMS, a complex and flexible treatment approach is favoured (1).

Concerning complementary medicine, a systematic review and meta-analysis on the use of Hypericum perforatum in dentistry reports about one randomized placebo-controlled trial (RCT) concerning herbal treatment in patients with BMS (10). Pain relief was not significant, but number of painful sites was reduced significantly. Further, there exists a Master's thesis analysing a homeopathic approach, Arsenicum album (Ars-a) 6X daily, for symptom management of BMS in a RCT. In this study 31 patients with different amount of comorbidities and different duration of symptoms were evaluated. After 90 days, oral burning in the verum group (n=16) improved significantly when compared to placebo (n=15). However, the symptoms persisted in 28 patients and concomitant symptoms were not addressed by the use of Arsenicum album 6X (11). In a second, open phase the patients received individualized Homeopathy (iHOM) and experienced a global improvement, including associated symptoms, such as depression, anxiety or gastrointestinal disorders. Considering this, it seems reasonable to present and discuss a detailed case report of long-lasting BMS using iHOM, an approach so far unestablished in the treatment for BMS.

PATIENT INFORMATION

The female, 38 year old patient was treated at the Institute of Complementary and Integrative Medicine (IKIM) University of Bern, Switzerland. In July 2014 she was referred by the department of aneasthesiology and pain medicine, Bern University Hospital where a BMS was diagnosed three years ago. Differential diagnoses were excluded from the referring clinic. At time of consultation, other concomitant symptoms were a moderate depression (assessed by the Beck Inventory Depression score), dysmenorrhea, overweight (Body Mass Index 25–30 kg/m2) and unspecific muscular pains which were suspected for being fibromyalgia. At the first consultation she reported daily constant burning and/or numbness of the left side of her tongue and the back of her throat. She also complained of insomnia, fatigue, dysmenorrhea and chronic muscle pains. For BMS-symptoms, she had tried Alpha lipoic acid, Pregabalin, Gabapentin, Venlaflaxin and topical Clonazepam as well as capsaicin, buprenorphine injections in the superior cervical ganglion, acupuncture and removal of all amalgam without any result. Since the start of her symptoms, she was also visiting a psychiatrist on regular basis, but she felt that this had no effect. Currently she was taking 50mg Chlorprothixen (Truxal), Thiamine and two herbal products (a valerian and hop combi-preparation and St. John's wort). Besides, she received hormonal injections every three months for contraception and took pain killers for the muscle and tissue pains.

CLINICAL FINDINGS

The patient presented herself with slow and sluggish speech as well as reduced facial mimic and needed to suck lozenges all day long, as this gave her relieve for dysesthesia in her oral cavity. In the anamnesis it was found, that BMS started after the intake of several drugs she had received for a post-partum depression after the birth of the patients' son, who was, by that time, four years old. She reported that she needed to change the anti-depressive therapy many times due to side-effects and that she couldn't remember after what particular drug or when exactly the symptoms of BMS began. Apart from numbness and burning of the tongue, she complaint of a salty or bitter taste in her mouth, which was ameliorated while eating. She considered this the reason why she needed to eat constantly. She had problems falling asleep, felt tired and without motivation during the day ever since the delivery of her child. Thus, she had never been able to take care of her son herself, but needed to rely on her husband and neighbours. Moreover, she became very irritated by him and struck him, when she was alone with him. Therefore, the family needed a nursemaid in order to provide the child's safety. Since the birth of her son, she also lost all her libido and over the past four years, she never had sexual intercourse with her husband. Around three months before the consultation at IKIM, pains in all her extremities started and had been suggested for being fibromyalgia from her primary care doctor. They were better from physical exercise, but she mostly felt too tired

to exercise. The dysmenorrhea had been present since her menarche and responded sometimes to hormonal treatment.

THERAPEUTIC INTERVENTION

The doctor at IKIM, who treated the patient, is trained in general medicine and holds a diploma for iHOM, a term which refers to individually administered homeopathic prescriptions of a single remedy, selected after a process of matching the present symptoms of the patient with those described in the homeopathic medicines' textbook "Materia Medica" following the „Law of Similars" (12). For the treatment of this case only highly diluted and succussed, i.e. so-called potentized homeopathic remedies were used. In particular, 'C-Potencies', in which the original substance is diluted 1:100, followed by 10 succussions were prescribed in single doses of 1x5 globules sublingualy or in 500ml tab water to be sipped throughout the day (liquid dilution) and 'Q-Potencies', in which the original substance is diluted 1:50'000, followed by 2-10 succussions were prescribed in a dosage of 3 drops in 100ml tab water daily. The number of the potency (e.g. 30, 200 or 3, 5 respectively) indicates the number of repetitions of this process. Two pharmacies, either Spagyros® (13) or Schmidt-Nagel® (14), approved by the Swiss Agency for Therapeutic Products (Swissmedic), provided the homeopathic remedies. They were prepared following the instruction of the European Pharmacopoeia, Monograph 1038 and 2045 and the Homeopathic Pharmacopoeia (HAB 5.2.2-5.2.6). For the homeopathic evaluation, the totality of presenting symptoms was considered and the practitioner edition of Complete Dynamics repertory © (15) was used.

Jurisdiction of the Ethics Committee of the Canton Bern was obtained and informed consent was given by the patient. The reporting is consistent with the HOM-CASE guidelines for reporting cases with homeopathic interventions ((16); Supplementary file 1)

The initial evaluation of the case is shown in table 1. It pointed to the remedy Sepia, which was prescribed as a single dose of C200 potency (13). The patient was also instructed to start with a Q3-potency (13) of the same remedy, if there was no result after two weeks. Changes of potencies and remedies are described in the section on follow-up and outcomes. Following a method described by Kent, the C-potency was risen, when the desired effect subsided (17). Additionally, Q-potencies were given for the relief of local symptoms, which aggravated after the C-Potency (17; Chapter 36).

FOLLOW-UP AND OUTCOMES

During the observation time the patient was asked to answer subjective outcome-assessments forms (Measure Yourself Medical Outcome Profile – MYMOP; (18). They were revised and summarised for the case-documentation, along with the notes of the treating doctor. Additionally, the subject was asked to assess the treatment outcome retrospectively in a validated questionnaire on Outcome Related to Impact on Daily Living (ORIDL; (19) in November 2017. The English assessment forms were translated by the treating doctor into German for patients' convenience.

Follow-up consultations initially took place every three to six weeks and were expanded up to every five months later, corresponding to the clinical improvement. After a relapse, the frequency of the visits needed to be reduced for a while and is currently being increased again. An outline of the consultations along with the given treatment and the assessed symptoms (18) is shown in table 2. In detail, the patient reported in the beginning rather an increase of the BMS-symptoms, but she felt generally better: she had more energy and less muscular pains from the first dose onwards. There also was immediately a marked improvement in the relationship with her son: she didn't need to strike him anymore and two months after the start of iHOM she was able to take care about him alone. Further, she reduced the Chlorproxithen to half. As the symptoms of the BMS appeared worse, the Q3 potency was stopped, but this didn't help. When she had a relapse of the emotional symptoms, the homeopathic dilution was risen and Sepia C1000 (12; liquid dilution) in September 2014 and end of September 2014 Sepia Q5 (13) was started. End of October she reported a slight improvement of the burning mouth with persistent improve of the mental features. As no further change could be noted and the salty taste in the mouth had increased, an intermediate dose of Natrium muriaticum C200 (13) was prescribed in November. This remedy is known in the homeopathic textbooks for being a complement to Sepia. After a short improvement a relapse of all symptoms to the state of September occurred and the therapy was changed back to Sepia in January 2015. This time, C30 (13) was given repeatedly (first daily, then once per week) and burning and numbness diminished considerably. By October of the same year there was only minor burning left on some days of the week. By this time she had reduced Chlorproxithen to 2,5 mg daily and stopped the herbal treatment. The treatment was changed again to Q-potencies (13), which were raised every 3 weeks by two potencies. End of 2015 she did not need painkillers anymore and had weaned of all medication completely. She was now fully able to manage the household and performed well with her son. Alongside, she joined a program for losing weight and reported to have gained interest in live again. However, by the time the BMS was under control she developed unspecific vertigo (normal physical examinations) and since the beginning of iHOM her dysmenorrhea had become worse, so that she changed the anti-contraceptive therapy at several times. In march 2016, after she had changed to a certain anti-

baby pill, she had pronounced metrorrhagia along with increased fatique and vertigo. She was given Ammonium muriaticum Q5 (13), but this didn't bring any change. The metrorrhagia stopped after she changed back to a hormonal depot-injection and the homeopathic treatment with Sepia Q (13) was re-established. After the Q19-potency there was a relapse of symptoms of all planes and she was given one dose of Sepia C30 (14) again and was told to take Sepia Q15 once every time the BMS-symptoms were prominent. This, and an increase of the potency to C200 (14) as well as an attempt with the remedy Nux-vomica (13) did not bring any result. As all symptoms still matched the symptoms described for Sepia in the 'Materia Medica', she received Sepia XM (14; liquid dilution) in March 2017 followed by Lactose without any homeopathic substance (Placebo). Also, Sepia Q5 (13) was given to be taken once in case of a burning tongue. Symptoms slowly improved, so that in June 2017 again nearly all symptoms but the dysesthesia in the mouth were gone. She reported in September 2017, that she had been well and with less BMS-symptoms until end of august and then the complaints relapsed. Another dose of Sepia XM (14; liquid dilution) followed by placebo was prescribed. In November 2017 the patient was nearly free of any complaints. She also reported, that she and her husband have been intimate for the first time since the birth of her son.

In the ORIDL-evaluation (19) she rated major improvement for the main complaint, the overall coping with the problem and the general well-being. From November 2017 onwards, there was no further change in the treatment or the severity of complaints. Sometimes, when the patient is not occupied or when she gets angry, she still has some minor discomfort in the mouth. It responds, however, always to a dose of Sepia Q15 (13).

Additionally, the modified Naranjo-criteria were used in order to assess the likely-hood of the documented changes being associated with the homeopathic intervention, as suggested in the HOM-CASE guidelines (QUOTE). The agreed score was 10 of 13 points (Supplementary file 2).

DISCUSSION

There are various treatment options for BMS, but they yield robust results consisting in a significant reduction of pain-scores in meta-analysis (8). Concerning alternatives, such as homeopathic treatment, limited data suggests that the homeopathic remedy Ars-a 6X, taken on a daily basis, may help for symptom relief when compared to placebo, but the actual changes in pain scores were small (Chebel). The prognosis of this disease is variable and there exist cases of complete spontaneous regression (9). Further, BMS is often associated with psychological distress (7) and Sardella et a. found, that the patients with spontaneous regressions also experienced profound psychological changes (9). In the open phase of the mentioned study on Ars-a 6X, 28 patients seemed to undergo

such changes under the treatment with iHOM, which followed the randomized phase with Ars-a 6X. The here presented patient reported an improvement of mood, energy and drive as well as other mental factors as well. appeared prior to the amelioration of the complaints of BMS, an effect which is stated to be associated with iHOM (16). For being a single case report, no conclusions about the influence of iHOM on the psychological factors can be drawn. However, there are certain coincidences which make an effect from the homeopathic remedy likely: Firstly, Sepia, as a homeopathic remedy is known for post-partum psychosis, hormonal dysregulation with loss of libido, general weakness with amelioration by movement and aggressive behaviour towards the own children. All of the symptoms were present in this case, and most of them were changing throughout the evolution of the patient. Secondly, there was an aggravation of BMS-symptoms and dysmenorrhea along with an amelioration of behaviour and drive in the beginning of the treatment: a situation which is unlikely to occur spontaneously. Thirdly, every time when the patient reported a relapse of symptoms, the indicated homeopathic remedy was repeated or the potency was risen and the symptoms reduced consecutively. Further, sometimes the amelioration of the patient's symptoms only responded to the higher potency, but not after the attempt of a repetition of the same potency. On the other hand, there exist a number of confounders, which limit the confidence in the observed result. Firstly, spontaneous regression is not excluded, even though BMS-symptoms existed for more than two years. Secondly, iHOM as an additional intervention could have been psychologically supportive as such and the remedy may not have had a specific effect. Lastly, other life-events may also have contributed to the improvement. Still, in the Naranjo-assessment two aspects considered to be attributed to iHOM, 'Did symptoms improve in the opposite order of the development of the disease' and 'Did at least one of the aspects of 'Hering's law of cure' apply to the case', were present and the score was considerably high (Supplementary file 2).

Besides the slightly increased oral burning, we observed no undesirable effects of the prescribed remedy, which could be managed with a change of potency. This event can regarded as an homeopathic aggravation as classified by Stub et al. (20) and the latter were graduated grade I according to the common terminology criteria for adverse effects CTCAE in a meta-analysis (21, 22). Thus, iHOM did not have safety-concerns in this case.

The limited experience of the presented case and the observations from Chebel (11), iHOM seems to support multi-dimensional recovery. This global effect of iHOM was also observed in cancer patients by Frass and colleagues: patients randomized to additional iHOM experienced significant improvement of general well-being as well as pain when compared to a cohort without this additional treatment (23).

As BMS is often associated with psychological distress (7), results of two RCT's on depression (24, 25) may also support a possible effect of iHOM in the treatment of BMS. In these studies, iHOM was equivalent to Fluoxetin in moderate and severe depression.

To sum up, an interdisciplinary approach for the treatment of BMS is favoured (26) and iHOM should be evaluated as one additional part of such an approach (26). As presented in this case, it may be considered as a safe treatment-option in cases refractory to other therapies. If considered, the patient should be referred to a specialist for iHOM.

CONCLUSION

This case shows how iHOM integrates the completeness of symptoms and complaints of a patient in the treatment approach. Considering the multi-factorial aetiology of BMS, iHOM might be of value in cases where an association of psychological factors and the neuralgic complaints is likely.

List of abbreviations

Ars-a	Arsenicum album
BMS	Burning mouth syndrome
iHOM	Individualized homeopathy
IKIM	Institute of Complementary and Integrative Medicine, University of Bern
MYMOP	Measure Yourself Medical Outcome Profile – Questionaire
ORIDL	Outcome Related to Impact on Daily Living – Questionaire

Declarations

Acknowledgments

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Conflict of interest

None to declare

Availability of Data and Materials

Data of this report is confidential. Anonymised material can be obtained from the author.

Ethics approval and consent to participate

Jurisdiction of the Ethics Committee of the Canton Bern was obtained with the Request ID 2017-00848 after the agreement of the patient, that the coded case data will be summarized and evaluated.

Consent for publication

Informed consent for publication was given by the patient. The signed document is confidential.

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Authors' contributions

Dr. med. K. Gaertner is responsible for data documentation and the preparation of the manuscript. Dr.med. M. Frei-Erb supervised the first author and added to the manuscript.

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